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## Title Page

### Editorial: EVERY PARTICIPANT IS A PI. CITIZEN SCIENCE AND PARTICIPATORY GOVERNANCE IN POPULATION STUDIES

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**Key messages:**

Citizen science and participatory research have become hot topics in various areas of biomedical research.

Although citizen science and participatory practices have been used successfully in a few instances within epidemiological research, the role they could play in this field has not been explored sufficiently.

This editorial proposes a model for an innovative use of citizen science in epidemiology called ‘Every participant is a PI’ (EPPI).

EPPI involves the creation of a non-profit citizen association of research participants to contribute to, and govern, a repository of samples and data for epidemiological research.

In describing EPPI, this editorial aims to further a discussion about the potential of citizen science and participatory research for the field of epidemiology.

**Editorial**

# **EVERY PARTICIPANT IS A PI: CITIZEN SCIENCE AND PARTICIPATORY GOVERNANCE IN POPULATION STUDIES**

## *Background: Declining participation in population research*

The willingness of people to participate in population-based studies has been declining for decades and is likely to continue to do so (1,2). Recently, large-scale cohort studies even had to be aborted due to poor recruitment (3). Although the use of online tools can boost recruitment in some contexts, attrition remains a big problem for such studies (4,5).

This situation is deeply concerning for epidemiological research. The effects of poor recruitment and high attrition are poor statistical power and an exacerbation of selection bias, which in turn diminishes the representativeness of population-based samples. For example, the disproportionate reduction in participation of people with low socioeconomic status (SES) could

have significant impact on the validity of study results, since one of the most robust findings of contemporary epidemiology is the association between low SES and disease risks (6). Unfortunately, recruiting participants for population studies from marginal or vulnerable groups seems particularly difficult (3).

According to Galea and Tracy (7), there are four structural drivers of decline in research participation, all of which are difficult to address by those currently planning large and long-term cohort studies: an escalation of the numbers of studies conducted; public mistrust in research; studies have become more burdensome for participants; and a general decrease in volunteerism in Western countries.

#### *Can participatory methods help? Citizen science in population research*

Recent patient- or participant-centered governance approaches to biobanking, whilst mostly focussing on matching consent and data protection requirements to the practical realities of population-based research, could also be seen as initiatives that focus on engaging participants more directly to increase initial sign-up and decrease attrition (8). We suggest a bolder approach, namely the introduction of so-called *citizen science* or *participatory research strategies* into population research. This would involve the transfer of more control over research agenda setting as well as the usage and handling of data and samples to participants. It could facilitate more meaningful engagement of people with the information that they contribute to research projects and the project as a whole, and enhance the volume and persistence of participation in well-designed epidemiological and biomedical studies.

#### *What are citizen science and participatory research?*

Digital technologies have already made it much easier for people without professional training to contribute to research (9). So-called ‘citizen science’ practices can take many forms,

including data and sample collection, data coding and analysis, and problem solving (10). And indeed, a quickly growing number of online platforms facilitate and involve contributions from lay people, generating biomedical datasets or enrolling volunteers to help with data analysis (11).

‘Citizen science’ is a broad church. It has origins in various fields and so far, the term describes a still emerging, heterogeneous field encompassing a broad range of activities (12,13). This is also reflected in the fact that the terms ‘citizen science’, ‘participatory research’ as well as ‘patient-led research’ are often used synonymously (14). While there is no universal definition of the term, all initiatives associated with citizen science have one thing in common: They include participation from volunteers who are not professional researchers (15). Contributions from volunteers can range from data collection that individuals input into a project run by professional scientists, to serving as advisers in the design of clinical trials, to volunteers themselves developing and performing a study on a treatment for their own illness, for example via a patient social network (some of these may be non-profit, some for-profit, and, in turn, collaborate with, e.g. pharmaceutical companies) (11). In the absence of a sharp analytical category, for the purpose of this paper we use ‘citizen science’ as a broad label that includes all of these understandings. We use ‘participatory practices’ as a generic term to refer to contributions that people make within citizen science project.

In biomedicine so far, most citizen science projects are run by professional scientists and researchers, with lay participation in the form of voluntary data and sample donations, or financial contributions to data-rich projects (crowdfunding) (16). In these projects, decision-making and research agenda setting largely rest with the researchers running the projects (they could be described as ‘top-down’ citizen science projects). However, interest is growing in initiatives where lay volunteers and patients take a more active role and function as part-

ners in their own right in research initiatives, or truly lead on research direction and decision making ('bottom-up' citizen science projects).

Some scholars have criticised citizen science, citing worries that due to lack of formal training, results might be of lower quality, or that such initiatives are vulnerable to commercial influences and interests, leading to biased results (17, 18). Certainly, not all participatory practices are created equal (11), but if done well, they could indeed provide a tool to support increased and meaningful participation of members of the public with science and research (16). User-friendly online tools and platforms in particular have expanded the ways in which individuals can control their data, paving the way for participant-centred initiatives in biomedicine and easing the dialogue between participants and researchers (19). Industry and policy makers have also devoted significant attention to these phenomena (22, 21, 22, 23). The sharply growing number of citizen science platforms and projects over recent years alone indicates that it draws in individuals and populations that might heretofore have not been interested in research at all.

So far, the role that citizen science and participatory practices could play in epidemiology has received limited attention. This is despite the fact that there are some successful initiatives of volunteers contributing epidemiologically relevant data, for example to detect epidemics faster than traditional hospital-based monitoring and tracking systems. Notable cases include the European Influenzanet (24), the US-based Flu-near-you (25), and other 'digital epidemiology' projects that provide local and timely information about disease dynamics in populations (26,27). Other examples include the harvesting of qualitative epidemiological intelligence contained, for example, within community observations and traditional oral history in veterinary epidemiology (28). These projects rely on volunteer participation and provide proof of principle that lay contribution in epidemiology can have very meaningful results for, e.g.,

public health. However, in terms of participation, such projects appear to be mostly organised in a top-down fashion, with the contribution of participants largely restricted to providing those running the projects with data; decision-making agency and control over the data and their further usage, as well as the direction of research, remain fully with those running the projects.

We believe that a further step should be taken in participatory epidemiological research. Epidemiological datasets and biobanks offer an as of yet unexplored opportunity to incorporate citizen science initiatives that are far more bottom-up into epidemiology, with volunteers themselves actively driving and controlling research. In the following, we sketch out the idea of a participatory model for population research that could be able to provide individual control over personal data as well as impact on research agendas, create robust datasets for biomedical research, and potentially counteract the response decline discussed above, all at the same time: Every Participant is a Principle Investigator (EPPI).

#### *Every Participant is a PI (EPPI)*

We propose to trial the creation of a non-profit citizen association of voluntary participants to form a sample and data repository they have largely control over. The basic idea is similar to the idea of health data cooperatives (29) or open data repositories such as DNALand (30) or OpenHumans (31). However, the crucial difference to these lies in the governance structures. Unlike the other repositories, EPPI is not a repository where people simply deposit data from various sources. It is an initiative where the main reason it exists in the first place is so that people can join with the specific goal of creating a joint repository *together*. Participants would decide together what types of data would be collected, stored, and used, and by whom. To the best of our knowledge, this has never been done in epidemiology.

Recruitment for EPPI could proceed in the following way: After the selection of a first population-representative sample, all potential participants would be informed about their selection and invited to participate in the EPPI project, and to form an association. To maximize the response, the invitation would be accompanied by regional and social media activities. The exact number of participants invited and the legal form of the EPPI association would depend on local circumstances and initial study aims, but in any case, membership would involve donation of samples and data into the EPPI repository. Should the final study population not be sufficiently large enough to answer the study questions, the sample could be extended appropriately. All EPPI participants would be informed initially that in contrast to existing biobanks, data handling, research agenda setting, and governance would not be handed entirely over to a third party, such as a university or research institute, but that members would remain involved in all of these processes if they so desire. Every participant would retain access to their own ‘raw’ data as it was entered into relevant databases in the cohort (32) and be able to participate in data governance. They could also propose study questions and secondary data uses. The central decision making body would be a governance board consisting entirely of participants. This board would appoint an advisory board that would include scientific and clinical experts.

EPPI is underpinned by the idea of solidarity. Current understandings of solidarity frame it as practices that harness people’s willingness to help others (33). Such pro-social willingness to work towards a common goal has been shown empirically to be an important motivation to participate in cohort studies and other research endeavours (34,35). Current biobanking governance frameworks, however, are not regularly reflective of solidarity-based motivations in participants, focusing instead on protecting individuals’ autonomy and shielding them from the risks of research (such as violations of privacy and data protection) (19). In EPPI, the sol-



idaristic idea that participants engage in a common endeavour, joining an initiative in which they decide *together* what the direction of research and particular research goals should be, would be front and center and serve as a motivational incentive to participate. In this model, *individual* control of data at every step of the way, while one obvious feature, is not the overarching goal. Instead, the emphasis is on enhancing reciprocity between different actors involved in research (i.e. between research participants, and participants and researchers), and further deliberation and genuine participation in the sense that people who do want to engage with decision making can do so. Further, mechanisms to ensure that people are not left alone when something does go wrong as part of this communal endeavour would be available (e.g. harm mitigation funds, 19,33).

We believe harnessing pro-social motivations to participate in research in a more systematic way could counteract the effects of mistrust and decreasing interest in more ‘traditional’ forms of research mentioned above (7), and that EPPI would be a suitable form to test this hypothesis. We also think this idea is eminently suited to be explored in epidemiology: Unlike, clinical medicine, epidemiology addresses health issues at group and societal level, and an EPPI initiative would allow for direct interaction, reciprocity and mutual support between members of a particular group.

Practically, as described, an EPPI initiative could be recruited wholly afresh, as described, or it could tag onto existing cohorts. In any case, it would typically be initiated by researchers/biobankers (but in theory also by patients or other citizens with a specific idea). Participants of a large existing cohort, for example a large research biobank, could also be invited to form an association as described. The association could be formed early on, whilst actual sample deposition and testing etc. would depend on the capacity of the study centre and the study aims, and would likely happen over a period of time. At least in the beginning and in

view of the significant costs state of the art-biobanking involves, to get the ball rolling, facilities for sample and data storage and handling would have to be made available by an institution that is later contracted by the association as service provider.

If recruitment in the way described was successful enough to start an association and establish a governing body, researchers would be part of any advisory board that the governing body would appoint, but would not have any further powers of agenda setting. All significant decisions would be made by the governance board consisting of participants. Once the EPPI cohort recruited is large enough for research, the governing body would decide on future study topics; which additional facilities to involve in future research; whom to allow use of the data and to what end; how to utilise and disseminate research results; etc. Researchers all over the world could apply for data and biomaterial usage, and suggest specific kinds of data or samples being collected. The association would appear as main or co-applicant in project proposals, appear as co-author on publications, participate in meetings of research consortia that use data that are co-owned by members, and so on. An association has the key advantage that it would outlast single projects, thus being able to co-govern long-term longitudinal studies. Moreover such an association could tap into additional sources of funding – for instance crowdfunding, which is often combined with or part of citizen science initiatives (36,16).

### *EPPI governance*

This idea might sound very unusual in view of the way existing research cohorts are governed (for an overview of the differences between established governance models and EPPI, see Table 1). However, there are already templates and precedents for specific elements of such an initiative. Models are in place that allow for individual control over data (37), and there have been prominent examples of successful patient-led research (38). It has also been

shown that participants can make productive as well as consumptive use of research(39). For example, the platform PatientsLikeMe, a social network for patients whose business model includes selling patient data to companies has facilitated the initiation of clinical studies by patients, for instance in the case of a study on the effects of lithium on the progression of amyotrophic lateral sclerosis (40). The extent to which such initiatives put patients and other citizens in control of research can be debated, but there is little doubt that they illustrate the interest of participants to engage more directly with research that could usefully be exploited to counteract the disenchantment with more ‘traditional’ forms of research mentioned above.

So far, the two objectives of data control and impact on research agenda setting, which would be joined in EPPI initiatives, have rarely been pursued together. Participant-centred governance of health datasets and biobanks has been typically understood as an extension and deepening of individual control over personal data and biological material on the basis of concerns for the autonomy and safety of participants (37). While such forms of governance may sometimes have an impact on how research is conducted and whether participants can influence research questions, planning and executions on the basis of their values and interests, such impact is mostly a side-effect, and not the main aim, of such models. In addition, fostering collective decision-making and deliberation certainly does not play a role in such governance. Here, we propose to go beyond individual and autonomy-based solutions and instead employ a governance approach that explicitly aims at opening up spaces where participants can put forward their ideas, research questions and concerns and deliberate them together. This may require broadly extending the understanding of participants’ roles in research.

**XXX Table 1 here XXX**

Table 1. Differences between three governance approaches to data repositories

### *Precursors and potential advantages*

While the devolvment of control and agency in EPPI will probably be regarded as a radical intervention by many, to our mind EPPI is no more than the next logical step in the trend to engage and involve patients and citizens in biomedical research in more active ways – although admittedly, it would be a large step and one that has not yet been taken in epidemiology (see table 2 for relevant differences between previous types of participatory initiatives relevant in epidemiology, and EPPI).

The implementation of participant-led features in contemporary epidemiological research continues a long standing tradition of popular epidemiology (41), whereby “lay” participants engage with scientists to tackle local issues and/or issues of concerns of those communities whose needs are often overlooked by traditional forms of research agenda setting (42). It is also an experience very much in line with one important early meaning of “citizen science” (43) and “science shops”, which – rather than denying the saliency of the different types of expertise of formally qualified scientists and citizens – tried to open up spaces where citizens could voice their priorities, concerns, and ideas and channel them into the mechanisms of agenda setting of science.

**xxx Table2 here xxx**

However, EPPI could not only learn from other citizen science initiatives, including those in other fields. It could also benefit from another important precursor: the Patient and Public Involvement movement (PPI) (44, 45). Efforts to include the patient perspective more explicitly in research – e.g. in clinical trial planning – have been pursued for some time now, and

are well documented (46, 47). EPPI could learn from and be informed by these experiences. For example, patients' and citizens' roles in PPI are often framed or understood in terms of 'public consultation' (48). This has led to concerns that such forms of lay participation might pitch the 'public view' vs. the 'scientific view', rather than steering the research agenda in directions agreed upon by professional scientists and lay citizens alike (49). We argue that because volunteers would steer the direction of research very actively in EPPI, it could breach this dichotomy. Moreover, PPI has been shown to be most relevant at the design stage of research (50), and the governance body of EPPI that would decide on this stage, made up of participants, is a central element of our proposed idea.

We believe that an EPPI initiative that was informed by previous, related experiences but pushed ahead of these, chartering new waters, could help reverse the aforementioned trends that drive declining participation rates in population-based research. As mentioned, traditional volunteerism is declining, but new form of sociality are developing at the same time, and benefiting from communication technologies. These can and should be fruitfully exploited by health researchers. Sharing personal medical data for the sake of research (either for curiosity, concern for a particular medical condition that affects the donor, or pro-social reasons) is becoming common, and especially among communities of citizens who seek to understand and tinker with their genomic data (51, 52). The empowerment of participants intrinsic in an EPPI initiative may also help address mistrust in biomedical science, and avoid the feeling of participants that they are being experimented upon as they learn about, intervene and ultimately decide about the research process. If successful, an EPPI initiative could eventually motivate people to participate, promote recruitment and retention, and cut attrition. At the very least, such an expectation, seen against the background of the emerging field of citizen science and participatory research as a whole, is not unreasonable.

In a system where participants are also co-investigators, some ethical issues and problems would fade into the background, as participants would overall be less in need of protection against third parties with competing interests (researchers, companies etc.), being themselves part of the governing bodies of research (53). Moreover, as mentioned above, the debate around novel forms of informed consent has brought attention to the fact that consent can also be seen as a form of consent to an entire system of governance, rather than to any single research initiative (54). In view of these considerations, it is at least likely that the more robust participatory features of an EPPI initiative would impinge positively on the perceived legitimacy of research, although this would have to be tested empirically in future EPPI studies.

#### *Potential pitfalls and challenges of the EPPI model*

Like every new idea, EPPI comes with a number of potential drawbacks and challenges, some of which can be anticipated, whilst others will only emerge in the pilot phase of such a project, or even at later stages.

Concerns that lack of training or experiences would lower the quality of results we believe could be assuaged by having an advisory board comprised of epidemiologists, biobanking experts and other researchers with relevant expertise, and by running EPPI, at least initially, on established IT, data and sample management infrastructure of existing cohorts.

However, another issue is possibly more difficult to avoid: PPI scholars have pointed out that projects of this kind can be vulnerable to ‘hijacking’, for example from highly educated individuals with professional and managerial backgrounds (55). Moreover, in the case of EPPI, researchers – and even executives from companies interested in cohort data – could become members and try to access collective data sets for their own work, as well as try to gain influ-

ence over agenda setting and decision making as part of the governing body. Similarly, the EPPI project overall, or the governance board more specifically, could become dominated by small groups of volunteer participants, or individuals with particular interests, that are especially vocal and engaged. This could lead to potential tensions within the EPPI association, as well as to unrealistic study aims, for example if individuals with very rare illnesses came to dominate the discussion, e.g. by demanding a study to assess the prevalence or incidence of that disease.

These are serious concerns, but we believe they can be addressed. In order to avoid any form of hijacking, EPPI would require well-designed policies for membership conduct, which would likely include high transparency regarding a member's potential conflicts of interest. Strategies and mechanisms of mediation would also have to be developed and implemented as part of the governance structures right from the start. To develop effective policies to this effect jointly, within a governing body made up of participants, is certainly a challenge. On the other hand, hijacking is not a problem exclusive to EPPI, and there are many policy examples for disclosing conflicts of interest, medication, etc. that EPPI could learn from.

It could be assumed that because EPPI research – just as other forms of population-based research – is not organised around specific diseases, it could lack the enthusiasm and engagement that can be observed in some citizen science projects, where research is driven by patients with very immediate needs. This could mean that, particularly in societies where individualistic values are strong, EPPI initiatives would not encounter much interest.

There is no doubt that initiatives that aim to combat particular diseases and develop treatments will be attractive to patients with that illness. However, many current citizen science initiatives, some of which are quite substantive in terms of the number of those involved, do not focus on particular diseases. In fact, a substantial number of citizen science projects takes place outside of the biomedical realm, and they attract substantive numbers of participation

(14), for example in several of the projects hosted in the platform Zooniverse (56). Moreover, the emerging forms of medical participatory research on online social platforms, e.g., elucidate a desire of citizens to have more direct involvement with research in general. If done well, citizen science can thus respond to citizens' desire to be more engaged with and contribute to science. Moreover EPPI would, in addition to previous initiatives, devolve significant control over the whole research process to participants, and this could open up new areas of interest for the subset of citizens already interested in participatory and voluntary initiatives. Whether appealing to all these interests and motives would be sufficient to recruit broad enough samples for EPPI initiatives to flourish needs to be tested in practice.

Another potential challenge EPPI shares with established forms of population research is to engage participants from marginalised groups and with lower socioeconomic status. Current recruiting strategies struggle to overcome barriers to accessing population-based research, and there is reluctance to participate originating, for example, in distrust (57, 58). This is also a well-described problem for PPI initiatives, even where PPIs exercises exist that are explicitly designed to reach minorities (48).

An EPPI initiative starting with a recruiting drive in an established cohort could utilise all the emerging online recruiting strategies available, and this might already contribute to engaging minorities and groups that heretofore have not been interested in research and who dropped out, or declined participation, in existing cohort studies. In addition, however, and in contrast to more traditional population-based research, EPPI could address the distrust that arguably plays a role in the difficulties to engage marginalised groups, and this could improve participation from these groups, once EPPI recruited outside the pre-existing cohort. EPPI explicitly devolves control over the research to participants and this could motivate those usually distrustful of research endeavours to participate.



We believe that this expectation that EPPI initiatives could reach yet under-represented populations is reasonable (59). EPPI initiatives could also, with online recruitment strategies as well as with participant-led, oral recruitment within local communities, help overcome well-known literacy problems that traditional population-based research suffers from.

There are also obviously different kinds of costs that such an experiment would incur. Setting up infrastructures for inclusive governance in an EPPI initiative, and for people to have access to their own raw data, will require significant resources. However, there is currently significant interest by funders to support citizen science and participatory initiatives, and this could mean additional streams of resources for the early stages of such initiatives. And, if the EPPI experiment was successful and provided a ‘proof of principle’ for bottom-up epidemiological citizen science, it would not be unreasonable to expect that such projects could eventually become financially self-sustaining, or at least considerably cheaper. They could draw in funding from those interested in the materials and data the association has control over.

### *Conclusions*

We expect that there might be other criticisms that could be levelled against this idea, and indeed we urge colleagues in the field to raise any concerns we have not anticipated in this editorial, so that the idea of an EPPI pilot can be refined. It is our explicit aim, by offering the idea of EPPI to the epidemiology community, to start a discussion about the future potential of this type of bottom-up citizen science in epidemiology. We believe that this potential is indeed significant, and that citizen science applications and participatory research and governance strategies could lead into a novel area to explore for the field. And, in any case: if we do not experiment with such initiatives, we will never be able to assess their real benefit, viability, and drawbacks.

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